

A Case Report of Primary Isolated Extrahepatic Hydatid Cyst of the Thigh

Primer İzole Uyluk Yerleşimli Ekstrahepatik Kist Hidatik Olgu Sunumu

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ABSTRACT

Hydatid cysts can appear as surprise masses in various anatomical regions of the body. Thyroid, breast, musculoskeletal system can be counted as atypical localization areas. The thigh, gluteal region, and neck are the most common locations for primary isolated muscular hydatid cysts. This text discusses the diagnosis, imaging methods, treatment strategy, and patient management of a primary isolated left thigh hydatid cyst. A 32-year-old male patient presented to the outpatient clinic with a slowly growing mass on his left thigh. Physical examination revealed a well-circumscribed tender mass in the upper medial part of the left thigh that did not cause skin discoloration. Ultrasonography revealed a multiloculated complex cystic mass measuring 10x6x12 cm between the thigh subcutaneous tissue and muscle planes. A multiseptal cystic mass was discovered between the muscle planes in the anteromedial part of the left thigh using contrast-enhanced tomography. The total cyst excision was performed under spinal anesthesia on the patient. During the two-year follow-up, no recurrence was detected in the patient's physical examination or radiological imaging. Extrahepatic primary hydatid cysts are frequently found subcutaneously. Thigh, gluteal region, neck, and extremity localization is extremely uncommon. Total cyst excision is the most commonly used surgical treatment method for intramuscular hydatid cysts.

Keywords: Echinococcosis, hydatid cyst, hydatid disease, extrahepatic cyst, thigh

ÖZ

Kist hidatik vücutta farklı anatomik bölgelere yerleşip sürpriz kitleler olarak karşımıza çıkabilmektedir. Tiroid, meme, kas ve iskelet sistemi atipik yerleşim bölgesi olarak sayılabilir. Primer izole müsküler kist hidatik en sık uyluk, gluteal bölge ve boyunda görülür. Bu yazıda; primer izole sol uyluk yerleşimli kist hidatığın tanı, görüntüleme yöntemleri, tedavi stratejisi ve hasta yönetimi sunulmuştur. Otuz iki yaşında erkek hasta sol uyluğunda yavaş büyüyen kitle şikayeti ile polikliniğe başvurdu. Fizik muayenede sol uyluk üst mediyal kısmında deride renk değişikliğine yol açmayan, düzgün sınırlı hassas kitle saptandı. Ultrasonografide uylukta subkütan doku ve kas planları arasında 10x6 x12 cm boyutunda multiloküle kompleks kistik kitle saptandı. Kontrastlı tomografide sol uyluk anteromediyal kısmında kas planları arasında yerleşmiş multiseptal kistik kitle tespit edildi. Hastaya spinal anestezi altında total kist eksizyonu uygulandı. Hastanın iki yıllık takibinde fizik muayene ve radyolojik görüntülemelerinde nüks saptanmadı. Ekstrahepatik primer kist hidatik sıklıkla subkütan yerleşimlidir. Uyluk, gluteal bölge, boyun ve ekstremitte yerleşimi oldukça nadirdir. Kas içi kist hidatik cerrahi tedavisinde kistin total eksizyonu en sık kullanılan yöntemlerdir.

Anahtar Kelimeler: Ekinekok, kist hidatik, hidatik hastalık, ekstrahepatik kist, uyluk

Introduction

Hydatid disease (HD) or echinococcosis; it is an endemic, zoonotic, parasitic infection caused by *Echinococcus granulosus* (sch) seen in both developing and developed countries and is considered a serious public health problem (1). Although this parasitic disease can be seen in any part

of the body, the liver (55-70%) and then the lungs (18-35%) are the most frequently affected organs (1). The brain, ovaries, pancreas, thyroid gland, breast, gallbladder, and heart are organs where echinococcus is rarely located and have a lower incidence of 8-10% (2). The rate of infection in the musculoskeletal system and primary soft tissue is extremely rare (0.5-4.7%), even in endemic areas (2,3,4).



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The disease can be discovered incidentally, or the patient can come with the mass to the affected location. A lump in the form of a soft tissue tumor, such as a sarcoma, liposarcoma, or lipoma, might be found during a clinical examination. The first preferred imaging method after the examination is ultrasonography (USG) (1,2,3). In the USG, one can see daughter cysts, vesicles, septa, hydatid sand, floating membranes, and cyst membranes (5). Serology can help in the differential diagnosis of HD and other cystic diseases. Magnetic resonance imaging (MRI), which depicts the location of the cyst and its relationships to muscles and other structures, provides the best imaging data for muscular hydatid cysts (5,6). In this article, the diagnosis of a primary isolated left thigh hydatid cyst, imaging modalities, treatment strategy and patient management are discussed with a case report.

Case Report

A 32-year-old male presented to the hospital complaining of a slow-growing, painless mass in his left thigh. The size of swelling gradually increased in the following period. He was a shepherd during his childhood. The patient's medical history was unremarkable. A sensitive lump of around 10*15 cm was discovered during a physical examination of the left upper thigh. The skin on top was normal (Figure 1). In the rest of the body, there was no more apparent or observable swelling. The lesion was detected by ultrasound as a 10x6x12 cm thick walled, multiloculated, complicated cystic mass in the medial side of the upper left thigh. Laboratory tests, including a complete blood cell count, serum chemistries, a coagulation profile, C-reactive protein, and



Figure 1. Swelling at the anteromedial aspect of the left thigh (the arrow)

erythrocyte sedimentation rate, showed normal results. In the serological examination, the indirect hemagglutination test was detected as 1/640 (positive >1/128). MRI revealed a large, thick-walled cystic mass lesion of approximately 18.5*15.5*8.8 cm on the anterolateral side of the left thigh, the largest of which was 4.4 cm, accompanied by multiple cysts (Figure 2). No other organs were affected by hydatid cysts, according to contrast-enhanced computed tomography (CECT) of the chest and abdomen. While CECT of the abdomen and chest did not reveal any other organ hydatid cyst involvement, but multiseptal cystic mass localized in the anteromedial part of the left thigh, measuring approximately 110*108 mm (Figure 3). The patient was treated with albendazole (400) mg in a day for two weeks before surgery. The wound was protected with a sponge dipped in hypertonic saline (3%). After injecting hypertonic saline (3%) into the cyst, a 10-minute waiting was required. Under spinal anesthesia, oblique incision anteromedially was performed. After the skin incision, it was observed that the cyst ruptured into the subcutaneous fatty



Figure 2. A. Coronal T1-weighted MRI study of the thigh demonstrate a large cystic lesion containing the smaller round cysts (arrow), B. Coronal T2-weighted demonstrate a large cystic lesion containing smaller round cysts (arrow)
MRI: Magnetic resonance imaging



Figure 3. Large cystic lesion containing the small round cysts (computed tomography scan with contrast) A: Coronal, B: Oblique, C: Sagittal plane images

tissue and was self-limiting. Up to 750 mL of filthy, decaying material with daughter cysts was extracted and cystectomy was performed (Figure 4). Thorough wash was given all cavities with 10% povidone iodine. In addition to the primary closure of the incision, hemovac drains (16 Fr) were inserted. The recovery time went without incident. The drain was removed on the third day. On the fifth day, the patient was released. The physical examination was unremarkable at the 1st and 3rd month follow-ups of the patient, and no recurrence was detected in the USG. In the macroscopic examination of the specimen sent to pathology; 4-5 transparent colored walland sac-like specimens, the largest of which is 7*7*0.1 cm and the smaller one is 2*2*0.1 cm, were observed. In microscopy examination; in the examination of the section prepared from the sent material; eosinophilicacellular cuticular membrane and scolex structures were observed on the wall, covered with a germinal layer inside. The case was diagnosed as “hydatidcyst” with the present findings (Figure 5).

Conclusion

A serious health issue around the world, HD, also known as cystic echinococcosis, is an endemic parasitic disease brought on by the larval form of the echinococcus. *E.*

granulosus and *E. multilocularis* cause cystic Echinococcosis and alveolar Echinococcosis in humans (1,2,3). While primary hydatid cysts of the thigh or other uncommon anatomical places have been recorded in the literature, hydatid cysts typically afflict the liver and lungs (1,2,3,4). The pathophysiology of musculoskeletal hydatid cyst disease is not clear. The two possibilities that get the most attention are subcutaneous contamination directly through damaged skin or lymphatic channel dissemination. The thigh is the most common subcutaneous location, followed by the gluteal area, the neck area, and the limbs (3,7). The location, size, and pressure of the cyst's expansion determine how the muscular hydatid cyst typically presents, but it typically takes the shape of a slow-growing, painless mass. Cyst formation is hampered by muscular lactic acidosis and muscle contraction. Parasitic cysts tend to develop around the muscles of the neck, thigh and roots of the limbs where muscle activity is lower and vascularisation is greater (8). There are serological tests available to diagnose hydatid cysts, however a negative test result does not rule them out entirely (1,2,4). Particularly in the detection of cystic membranes, septa, and hydatid sand, floating membranes, daughter cysts, and vesicles, as well as in the illness diagnosis, USG has a high accuracy rate (5,7,9). MRI is the preferred examination in case of suspected intramuscular HD, as in all muscle-joint imaging. The differential diagnosis of cystic lesions is aided by the detection of multiloculated polycystic mass lesions on MRI as well as the presence of a two-layered wall of collagen tissue and vascularized pericyst (5,8,10). The preferred method of therapy at the moment is total surgical excision of the cyst, followed by extensive irrigation of the soft tissues around it with hypertonic saline to avoid recurrence. The following surgery, these operations should be accompanied with the administration of systemic antiparasitic medications. When cystic lesions are seen on the body, the differential

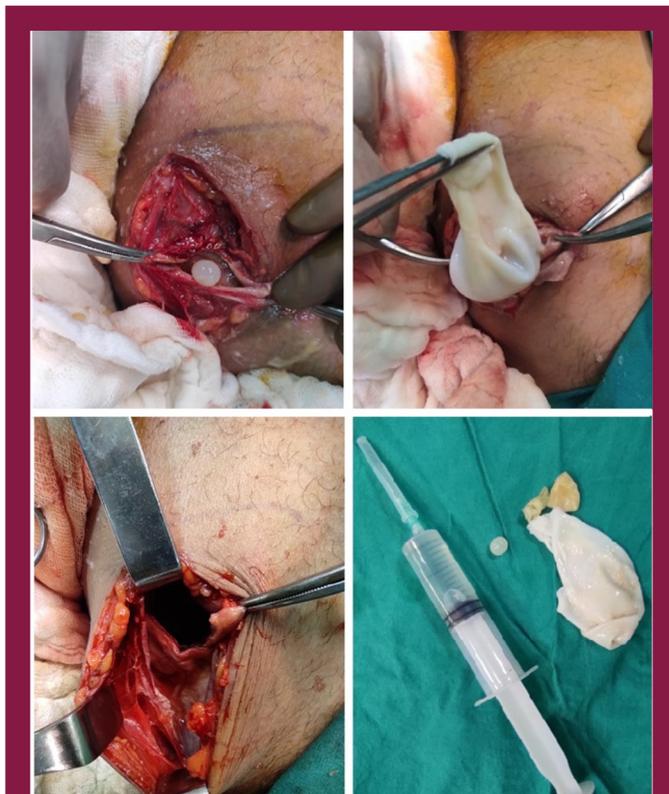


Figure 4. Peroperative images of thigh and specimens

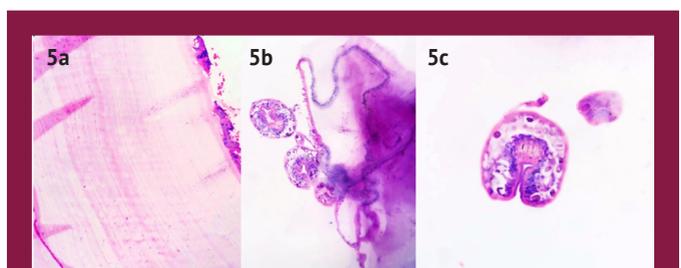


Figure 5. Pathology findings. **5a:** The appearance of eosinophilic stained, lamellar structure, acellular cuticle layer, and scolex structure in the cyst (HEX40), **5b:** The appearance of eosinophilic stained, lamellar membrane and associated scolex in the cyst (HEX20), **5c:** Scolex structures (HEX40)

diagnosis should include a hydatid cyst, especially in endemic populations. MRI is accepted as one of the best diagnostic techniques in the diagnosis and surgical planning of primary muscle localized hydatid cysts. Total excision of the cyst or pericystectomy is the most commonly used methods in the surgical treatment of intramuscular HD.

Information

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Ethics

Informed Consent: Written informed consent was obtained from the patients for the publication of this case report.

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Authorship Contributions

Surgical and Medical Practices: R.S.A., M.Ç., S.D., Ş.K., Concept: R.S.A., S.D., Ş.K., Design: R.S.A., M.Ç., Data Collection or Processing: R.S.A., S.D., Ş.K., Analysis or Interpretation: R.S.A., Ş.K., Literature Search: R.S.A., Writing: R.S.A., S.D., Ş.K.

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